



I'M SLEEPY: A short pediatric sleep apnea questionnaire



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ABSTRACT

Background: Pediatric obstructive sleep apnea (OSA) is a prevalent but under-diagnosed disease. The importance of screening for OSA has been emphasized in the recently published guidelines for the diagnosis and management of childhood OSA. Several pediatric OSA questionnaires are available, but are complicated to use or not sensitive enough for screening.

Methods: In this study we developed an 8-item (I'M SLEEPY) screening tool for pediatric OSA. One hundred and fifty children referred for evaluation at a pediatric sleep clinic and their parents completed the questionnaire and had a polysomnography. Two further questionnaires were developed: I SLEEPY and I'M SLEEPY versions. The questionnaires' scores were compared to the apnea hypopnea index (AHI) and the validity of each questionnaire was evaluated.

Results: The I'M SLEEPY version was found to have the highest sensitivity (82%) and a modest specificity (50%) for OSA diagnosis.

Conclusion: I'M SLEEPY is a sensitive and easy-to-use screening tool for pediatric OSA. It is intended to be used by the primary physician in every suspected case of OSA. Larger studies are needed in the primary care setting for the validation of this tool.

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1. Introduction

Pediatric obstructive sleep apnea (OSA) is a common disorder, affecting 1–4% of the general pediatric population [1,11,14]. It manifests as snoring, labored breathing and apnea during sleep and has significant adverse effects on the child's daytime behavior, performance and health, including daytime sleepiness, school performance deterioration, neuro-cognitive impairment, failure to thrive, hypertension, cor pulmonale, pulmonary hypertension and even death [1,14]. However, pediatric OSA is an under-diagnosed disease. The gold standard for diagnosis is an overnight polysomnographic study (PSG), but this test is expensive and, apart from large urban centers, has relatively low availability internationally, particularly in the pediatric population. Recently, specific guidelines for the diagnosis and management of pediatric OSA have been published, stressing the importance of screening for snoring and other OSA symptoms in every child [12]. In order to correctly identify

children with possible OSA who should be referred for a sleep study, a number of questionnaires have been developed [5,17]. One of the most accepted is the Sleep Related Breathing Disorders (SRBD) Questionnaire, which is a 22-item questionnaire, developed by Chervin and colleagues. However, these questionnaires are, in general, either too long to be used as a screening tool by the pediatrician or family physician (with 22–40 questions each) or do not have high sensitivity [3,7]. A short and sensitive screening tool is urgently needed in order to facilitate the accurate triage of patients with potential pediatric OSA, increase the rate of pediatric OSA diagnosis and prevent the long-term complications of the disorder. We have recently developed such a scale for the detection of adult OSA (STOP-Bang), which has become widely used [6].

In this study we created and validated an 8-item OSA pediatric questionnaire in our cohort. This questionnaire will be helpful in screening for pediatric OSA and facilitate the decision as to which patient should be referred for a PSG for further evaluation.

2. Methods

2.1. Study population

The study was conducted in the Youthdale Child and Adolescent Sleep Clinic, Toronto, Ontario, Canada. Ethic approval was obtained

Abbreviations: AHI, apnea hypopnea index; BMI, body mass index; OSA, obstructive sleep apnea; PSG, polysomnographic study.

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from the Institutional Review Board Services (Aurora, Canada). Children aged 3–18 years, who were referred to the clinic for a sleep evaluation, were included in the study. Children older than 7 years who were unwilling or unable to give assent, children who were already diagnosed with OSA, or children who were not eligible for a sleep study (e.g., had a sleep study during the previous two years) were excluded. All the children who visited the clinic and met the inclusion criteria were approached by the study staff. If informed consent was obtained, the parents/guardians were asked to complete an 8-item questionnaire. Children aged 7 years and older were asked to complete a pediatric version of the same questionnaire before undergoing the sleep study.

2.2. Development of the IF SLEEPY questionnaire

In order to choose the most important questions for the diagnosis of pediatric sleep apnea, a three-step approach was utilized. The first step was a semi-expert opinion – a group of sleep specialists, sleep technicians and researchers gathered and wrote 17 questions intended to diagnose pediatric OSA (Appendix 1). The questions were designed in a yes/no format for simplicity of completion and scoring. The second step was the utilization of an expert opinion: leading international pediatric sleep specialists were approached by email and asked to rate the 5 most important questions out of the 17, and to add more questions if they so wished. The 8 most frequently chosen questions created the IF SLEEPY questionnaire: I – Is your child often Irritated or angry during the day?; F – Does your child often Fidget and/or is hyperactive?; S – Does your child usually Snore?; L – Does your child sometimes have Labored breathing at night?; E – Ever noticed a stop in your child's breathing at night?; E – Does your child have Enlarged tonsils and/or adenoids?; P – Does your child have Problems with concentration?; Y – Does your child often Yawn or is often tired/sleepy during the day?.

The questions were also modified for children: I – Are you angry a lot?; F – Is it difficult for you to sit quietly? Do you feel that you always have to be “on the move”?; S – Do you snore at night?; L – Did your parents or a friend tell you that your breathing is “difficult” at night?; E – Did your parents or a friend tell you that you stop breathing at night?; E – Do you have problems with your tonsils or adenoids (glands inside your mouth)?; P – Is it difficult for you to focus (at school or at home)?; Y – Do you feel tired or sleepy a lot?.

2.3. Definitions

The score of each questionnaire was calculated by adding the positive responses. The parent and child versions were scored independently. A child at high risk for OSA was defined as a child with a score of ≥ 3 .

OSA was defined as apnea hypopnea index (AHI) ≥ 1.5 /h. Mild OSA was defined as AHI ≥ 1.5 and < 5 /h, moderate as AHI ≥ 5 and < 10 /h and severe as AHI ≥ 10 /h [13].

Overweight/obesity were determined by calculating the body mass index (BMI) percentile. The BMI percentile is the percentile ranking of each child based on a comparison of their BMI with that of other children of their same age and gender as obtained from the Centers of Disease Control (CDC). Overweight is defined as BMI percentile ≥ 85 and < 95 , and obesity is defined as a BMI percentile of 95% or higher, according to the CDC definitions [3].

2.4. Validation of the questionnaire

The questionnaire was separately completed by a parent/guardian and children older than 7 years (the child version) before undergoing PSG. Children older than 7 years, with their parents

help, were also asked to complete a pediatric-modified STOP-Bang questionnaire (Appendix 2), without the 6th question (i.e., age > 50 years). The responses of the children and their parents/guardians were gathered in an Excel sheet (Office 2007, Microsoft), together with epidemiological (e.g., age, sex) and clinical (e.g., height, weight, BMI) data.

2.5. Sleep study

A one night, in-laboratory PSG was conducted for every child by a pediatric sleep technologists. Each study lasted 8–10 h and included overnight monitoring of electroencephalogram, electro-oculogram, EKG, chin and anterior tibial electromyogram, nasal pressure transducer, oral thermistor, a snore sensor, respiratory inductive plethysmography, pulse oxymetry, end-tidal CO₂ monitors and continuous video monitoring. The studies were conducted and scored according to the American Academy of Sleep Medicine manual for the scoring of sleep and associated events [9] with a computerized system (XL-TEK, Oakville, Ontario, Canada). Each epoch of every study was initially scored by the software and then manually by an experienced pediatric sleep technician. The study reports were assessed and approved by a sleep physician. The staff performing, scoring and approving the PSG study and report were blinded to the score on the questionnaire. Data from the study report (i.e., sleep architecture, AHI, respiratory disturbance index, minimal O₂ saturation and CO₂ values) was added to the excel sheet.

2.6. Sample size determination

It has been suggested that meaningful qualitative conclusions can be drawn from receiver operator characteristic (ROC) experiments performed with a total of about 100 observations (Metz 1978). Given this rule of thumb, in combination with an estimated attrition rate of 10% and incomplete data and technical difficulties in 20%, we enrolled 150 patients.

2.7. Statistical analysis

Differences between patients with and without OSA diagnosis were assessed using Student's *t*-test (for BMI percentiles, sleep onset latencies and arousal indexes) and the chi-square tests (for male:female ratios). A *p* value ≤ 0.05 was considered significant. The sensitivity, specificity and positive and negative predictive values (PPV and NPV) of the study questionnaires in diagnosing pediatric OSA were assessed via ROC curves (SPSS software), based on the AHI as a gold standard. The area under the curve (AUC) was calculated. This procedure was done separately for the parents'/guardians' responses and for the childrens' responses.

3. Results

3.1. Population

One hundred and eighty five children and their parents gave consent to participate in the study. Twenty five children were excluded (23 had a previous diagnosis of and treatment for OSA and two were older than 18 years), and 150 children were included in the study. The demographic characteristics of the children are shown in Table 1.

3.2. PSG results

Forty-nine out of 150 children (33%) were diagnosed with OSA. Twenty-five (51%) were diagnosed with mild, 9 (18%) with moderate and 15 (31%) with severe OSA.

Table 1

Epidemiological and clinical characteristics of the patients.

	All patients	OSA	No. OSA	Significance
No. of patients	150	49	101	
Age	11.4 ± 4.1 years	10.9 ± 4.5 years	11.7 ± 3.8 years	ns
Male:female ratio	2:1	3.4:1	1.7:1	
BMI (kg m ²)	21.1 ± 6.4	24.1 ± 8.4	19.7 ± 4.6	p = 0.001
BMI percentile	66.0 ± 31.3	75.8 ± 31.7	61.3 ± 30.1	p = 0.009
Neck circumference (cm)	31.0 ± 4.8	31.9 ± 6.2	30.6 ± 3.9	ns
Sleep onset latency (min)	33.8 ± 33.0	29.5 ± 25.8	35.8 ± 35.9	ns
Sleep efficiency (%)	82.6 ± 11.3	82.2 ± 8.5	82.8 ± 12.5	ns
REM latency (min)	138.7 ± 77.2	141.3 ± 77.4	137.5 ± 77.5	ns
Stage 1 (%)	5.0 ± 4.0	6.4 ± 6.0	4.3 ± 2.3	p = 0.02
Stage 2 (%)	43.5 ± 9.1	42.2 ± 9.5	44.2 ± 9.0	ns
SWS (%)	22.6 ± 6.9	22.0 ± 7.8	22.9 ± 6.4	ns
REM (%)	18.8 ± 7.7	18.9 ± 10.1	18.7 ± 6.3	ns
Arousal index	13.2 ± 8.1	17.9 ± 11.4	10.8 ± 4.3	p < 0.001
AHI	4.2 ± 11.4	11.8 ± 17.7	0.5 ± 0.4	p < 0.001
Minimum O ₂ saturation	90.7 ± 5.0	88.3 ± 7.3	91.9 ± 2.8	p = 0.001
CAI	1.0 ± 2.3	1.5 ± 3.6	0.76 ± 1.1	ns
PLMI	1.1 ± 3.0	1.5 ± 3.8	0.9 ± 2.6	ns

All values above are expressed as mean ± SD.

BMI: body mass index; SWS: slow wave sleep; AHI: apnea hypopnea index; CAI: central apnea index; PLMI: periodic limb movements index.

Table 2

Validity of the study questionnaires.

	Parent			Child			STOP-BNG
	IF SLEEPY	I SLEEPY	I'M SLEEPY	IF SLEEPY	I SLEEPY	I'M SLEEPY	
Respondents (n%)	137 (91%)	137 (91%)	137 (91%)	115 (77%)	115 (77%)	115 (77%)	69 (46%)
Positive questionnaire response ^a (n%)	99 (72%)	82 (60%)	91 (66%)	70 (61%)	56 (49%)	65 (47%)	16 (23%)
Positive questionnaire response plus AHI ≥ 1.5 (n%)	38 (25%)	37 (25%)	50 (33%)	22 (15%)	19 (13%)	59 (39%)	6 (4%)
Sensitivity (%)	78%	76%	82%	45%	39%	47%	12%
Specificity (%)	40%	55%	50%	52%	63%	58%	90%
Positive predictive value (%)	38%	45%	44%	31%	34%	35%	38%
Negative predictive value (%)	78%	82%	85%	66%	68%	69%	68%
% False positives	60%	45%	18%	48%	37%	53%	10%
% False negatives	22%	24%	50%	55%	61%	42%	88%

^a Those endorsing three or more symptoms or complaints on the questionnaires. The percentages in brackets are out of 150 children participating in the study.

Twenty-nine patients (19%) were overweight (BMI percentile ≥85%) and 33 (22%) were obese (BMI percentile ≥95th). The mean BMI percentile was higher among children with OSA ($p = 0.009$) as compared to those without an OSA diagnosis. The arousal index ($p < 0.001$) and percentage of stage 1 sleep ($p = 0.002$) were significantly greater and the minimum nocturnal oxygen saturation ($p = 0.001$) significantly lower among children with OSA. No other statistically significant differences in sleep study parameters were noted between the study groups.

3.3. IF SLEEPY

One hundred and thirty seven parents/guardians completed the IF SLEEPY questionnaire (91% response rate). On the parent IF SLEEPY, 99 (72%) children had a score ≥3 and were considered at high risk for OSA. Thirty eight children with high risk had OSA, resulting in a sensitivity of 78% and a specificity of 40%. Other validity indices are listed in Table 2.

One hundred and fifteen (77%) children completed the child IF SLEEPY questionnaire and 70 children scored at high risk for OSA (≥3). Twenty two children with high risk had OSA. The sensitivity and specificity of the child IF SLEEPY for OSA diagnosis were 45% and 52%, respectively.

3.4. I SLEEPY

In order to improve the specificity of the IF SLEEPY, we modified the scale by erasing the F question (“does your child fidgets/is he

hyperactive?”), as symptoms of hyperactivity are common but not specific to OSA, thus creating the I SLEEPY version. The sensitivity and specificity of the *parent* I SLEEPY questionnaire in diagnosing OSA were 76% and 55%, and the sensitivity and specificity of the *child* I SLEEPY were 39% and 63%, respectively.

3.5. I'M SLEEPY

In order to improve sensitivity, a parameter of overweight/obesity was added to the scale, thus creating the I'M SLEEPY version, where the “M” stands for body Mass index (Table 3). Every child with a BMI percentile of 85% or higher received an additional point to the I SLEEPY scale score, and the sensitivity and specificity were calculated: for the parent version 82% and 50% and for the child version 47% and 58%, respectively.

3.6. STOP-BNG

Sixty nine children (46%, 7–18 years of age) and their parents completed the modified STOP-Bang questionnaire. The sensitivity and specificity of the modified STOP-Bang for pediatric OSA diagnosis was 12% and 90%, respectively.

4. Discussion

Pediatric OSA has significant health implications, including sleep disruption, daytime sleepiness and fatigue, cognitive and behavioral effects and growth retardation [1,14]. In this study, the

Table 3
I'M SLEEPY questionnaire.

Parent version:
I – Is your child often <u>I</u> rritated or angry during the day?
M – Body <u>M</u> ass index above 85%?
S – Does your child usually <u>S</u> nore?
L – Does your child sometimes have <u>L</u> abored breathing at night?
E – <u>E</u> ver noticed a stop in your child's breathing at night?
E – Does your child have <u>E</u> nlarged tonsils and/or adenoids?
P – Does your child have <u>P</u> roblems with concentration?
Y – Does your child often <u>Y</u> awn or is often tired/sleepy during the day?
Child version:
I – Are you angry a lot?
M – Filled in by the doctor: body mass index above 85%?
S – Do you snore at night?
L – Did your parents or a friend tell you that your breathing is "difficult" at night?
E – Did your parents or a friend tell you that you stop breathing at night?
E – Do you have problems with your tonsils or adenoids (glands inside your mouth)?
P – Is it difficult for you to focus (at school or at home)?
Y – Do you feel tired or sleepy a lot?

I'M SLEEPY questionnaire was developed and validated as a screening tool for pediatric OSA. This questionnaire includes 8 yes/no questions and was found to be sensitive (82%), but less specific (50%) for the diagnosis of OSA. Further, this study compared questionnaire responses of both parents and children and noted that parental reports were more reliable.

Several screening tools for pediatric OSA are available in the literature. One of the first to be published is the Brouillette score [4]. This score is calculated according to the formula: $1.42D + 1.41A + 0.71S - 3.83$, with A for observed apnea, D for dyspnea and S for snoring. A score of <-1 is indicative of no OSA and a score >3.5 is indicative of OSA. The Brouillette score was initially found to have high sensitivity but low specificity for OSA. Subsequent studies questioned its utility in different patient populations [2,16]. Another questionnaire for the diagnosis of pediatric OSA is the OSA-18, an 18-items questionnaire focused on sleep disruption, physical and emotional distress and diurnal symptoms [8]. The scale had significant relationship with the Respiratory Distress Index (RDI), but was later found to be only 40% sensitive in diagnosing pediatric OSA [3].

A well-accepted pediatric OSA questionnaire is the Sleep-related breathing disorder Scale (SRBD scale), a subscale of the Pediatric sleep questionnaire (PSQ). This scale was developed by Chervin and has 85% sensitivity and 81% specificity for the diagnosis of OSA [6]. It includes 22 items of nocturnal (mainly respiratory) and diurnal symptoms, growth and attention deficit/hyperactivity symptoms and takes several minutes to complete. A positive response for more than 7 items indicates possible OSA. Owens and colleagues developed the Children's sleep habits questionnaire (CSHQ): a 45 items scale which provides a total and eight sleep domains sub-scores, including sleep-disordered breathing [15]. A cutoff score of 41 yields a sensitivity of 80% and specificity of 72% for the diagnosis of sleep disorders. And last, the sleep disorders inventory for students (SDIS) is another questionnaire for several sleep disorders in children and adolescents, with a subscale for OSA [10]. It was found to have good sensitivity (86–91%) and specificity (62–88%) for diagnosing OSA and includes 40 items. A major drawback of the above 3 screening tools is the large number of items which render them impractical for routine clinical use, especially in a pediatric clinic.

In conclusion, several pediatric OSA screening questionnaires are available and some of them have good sensitivity and specificity for the diagnosis of OSA. However, due to their complexity of completion, they are mainly used for research or

in sleep clinics, and not for screening of children in the primary care clinic. The short questionnaire developed in this study is better suited for the primary care setting: it takes less than 1 min to complete and the catchy mnemonic makes it easy for the family physician, pediatrician, ENT surgeon and dentist to memorize and use when the question of OSA arises in relation to their patients.

Of the questionnaires tested in this study, the parent versions had overall higher sensitivities and specificities compared to the children's self-report version. The parent version of the I'M SLEEPY questionnaire had the highest sensitivity (82%) and negative predictive value (85%) but a modest specificity (50%). The strength of the I'M SLEEPY questionnaire would be to help the primary care physician in identifying those children who are more likely to have OSA and also to remind the physician as to which questions are most important to ask when suspecting that their pediatric patients may have OSA.

The STOP-Bang screening questionnaire for sleep apnea has been shown to reliably detect sleep apnea in adults [8] but was found to have a very low sensitivity in the pediatric population, even after removal of the 'age >50 years' item. The differing etiologies and clinical presentations of OSA in children and adults probably accounts for this disparity in the performance of the STOP-Bang [11]. This finding underscores the need for the development of reliable screening questionnaires specifically targeted for the pediatric population.

There are several limitations to the study. The first is the low overall specificities of the IF SLEEPY, I SLEEPY and I'M SLEEPY questionnaires in diagnosing pediatric OSA. As a screening tool, it means that approximately 2 children will be referred for a sleep study for every single diagnosis of OSA. As OSA has significant negative physical, mental and academic consequences, and is readily treatable, we believe the cost-benefit ratio is acceptable.

The second limitation is the relatively small number of patients and the single center design of the study. The third limitation is a sample bias. The questionnaire was completed by patients referred for a sleep study, as opposed to primary care patients. We are now conducting an additional study in order to validate the questionnaire in general pediatric clinics.

5. Conclusion

The I'M SLEEPY questionnaire is a screening tool for OSA in children with a high sensitivity, that is, it is accurate in identifying which children are at high risk of having OSA. It is intended to be used in primary-care clinics in every suspected case of pediatric OSA. Further studies are needed in order to test this tool in larger, primary care settings.

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Conflict of interest

The authors have no conflicts of interest to disclose.

Author contribution

Gili Kadmon – primary responsibility for the study design, data collection and analysis, and manuscript drafting and revision.

Sharon Chung – study design, data collection and analysis and manuscript revision.

Colin Shapiro – study design, data collection and manuscript revision.

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Appendix 1

Questions written by the semi-expert opinion panel

1. Does your child snore at night?
2. Does your child have labored breathing at night?
3. Does your child have breathing stops at night?
4. Does your child breath with his/her mouth open?
5. Does your child complain of dry mouth in the morning?
6. Does your child ask for a drink immediately after waking up?
7. Does your child wet the bed occasionally?
8. Is your child sleepy or tired during the day?
9. Does your child complain of headaches in the morning?
10. Is your child grumpy in the morning?
11. Is your child overweight?
12. Has your child stopped growing at a normal rate?
13. Is your child significantly inattentive? (easily distracted, does not seem to listen, has difficulties with organizing)
14. Is your child hyperactive? (fidgets with hands, "on the go", squirms in seat, interrupts on others)
15. Does your child have enlarged adenoids and/or tonsils?
16. Does your child have an overbite?
17. Does your child have developmental delay or genetic malformation?

Appendix 2

The pediatric STOP-BNG questionnaire:

- S – do you Snore?
 T – do you feel Tired, fatigued or sleepy during the day?
 O – has anyone Observed you stop breathing in your sleep?
 P – do you have high blood Pressure?

B – BMI > 35?

N – Neck circumference >40 cm/15.7"?

G – Gender = male?

(the A – Age >50 years? Question used in the adult version is omitted for children).

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